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Case Report

Pre-operative Occurrence of First Bite Syndrome in Two Cases of Parotid Gland Tumour

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Abstract

Introduction: First bite syndrome (FBS) is a gustatory-evoked painful condition that is characterised by the onset of severe electric shock-like pain in the periauricular region. In the majority of patients, FBS develops postoperatively. However, in rare instances, it may present in a pre-operative setting. **Case Description:** Two cases of FBS developing preoperatively secondary to parotid gland tumour are presented. The patients, 54 and 30 years old, presented with complaints of electric shock-like pain localised over the periauricular region. In each case, the pain was triggered following the first bite of the meal. In both cases, the pain lasted for few seconds and with subsequent bites subsided only to return at the beginning of the next meal. **Practical Implications:** Pre-operative occurrence of FBS in the absence of a history of trauma or surgery should necessitate a thorough investigation and follow-up for an underlying salivary gland tumour.

Key words: Cancer pain, first bite syndrome, neuralgia, parotid neoplasms

Introduction

First bite syndrome (FBS) or gustatory neuralgia is a gustatory-evoked painful condition that is characterised by excruciating and lancinating electric shock-like pain, lasting for seconds, in the periauricular region.[1,2] The pain is triggered during the initial bites of a meal and alleviates with subsequent masticatory movements. There are no reports of pain in between meals. However, sometimes, the pain may be triggered after thinking about food. The pain may present in association with severe cramping or mild sweating in the region.[1-3] This is a separate entity from Frey syndrome (auriculotemporal syndrome or gustatory sweating) which is characterised by sweating and flushing in the cutaneous distribution of the auriculotemporal nerve following a gustatory stimulus. In Frey syndrome pain is an uncommon finding (<10%). However, if present it is described as a mild constant dull ache or burning sensation.[1]

The vast majority (~95%) of patients develop FBS after experiencing trauma in the head-and-neck region or postoperatively following resection of the sympathetic chain or parotid gland or after surgery involving parapharyngeal space, infratemporal fossa, carotid bifurcation or internal carotid artery.[4,5] The incidence of FBS developing postoperatively in patients undergoing surgery of the deep parotid gland, parapharyngeal space and infratemporal fossa is estimated to be 9.6%.[5] However, there are only a handful of cases that have been reported of FBS, preoperatively. These have been associated with the presence of tumours in the parotid or submandibular glands or parapharyngeal region.[6-9]

Salivary gland tumours constitute about 5–6% of head-and-neck cancers.[10] The prevalence and incidence of salivary gland tumours are 4.7 and 0.9 cases/100,000 individuals, respectively.[11] Majority (around 80–85%) of these tumours are benign and affect the parotid gland. Benign tumours are more common in females. However, malignant tumours affect males and females equally.[11] The clinical features of parotid gland tumour consist of painless swelling, loss of the function of the cutaneous branches of the facial nerve, numbness or paraesthesia and tenderness in the region of the gland.[11-13]

This article presents two case reports of patients that developed FBS preoperatively secondary to parotid gland carcinoma.

Case Report

Case I

A 54-year-old male presented with an 8-month history of electric shock-like lancinating pain localised over the right side preauricular and posterior mandibular regions. The pain was severe (10/10) in intensity and would be triggered following the first bite of a meal. This would continue for the initial few bites and subsequently subside. In addition to electric pain, the patient complained of a constant dull ache in the region. The dull pain was mild (2/10) in intensity and did not vary with masticatory movements. There were no reports of pain in between meals. Three weeks before the appointment, the patient developed a soft non-tender swelling over the right side posterior mandibular region and weakness in the right side lower lip movements.

Medical history of this patient was unremarkable. Examination of the head-and-neck region was significant for a 20–25 mm nodular soft swelling in the right side posterior mandibular region. There was mild tenderness (1/10) to palpation in the right posterior mandibular and submandibular regions. Cranial nerve examination revealed mild weakness in the function of the right side marginal mandibular branch of the facial nerve. The remainder of the examination was unremarkable. A diagnosis was made and the patient was referred for computed tomography (CT) of the head-and-neck region and fine-needle aspiration (FNA) of the nodular swelling.

The CT revealed an approximately 2.1 cm × 1.9 cm × 3.6 cm heterogeneously enhancing mass with speculated margins at the junction of the superficial and the deep lobe of the right parotid gland. The mass had completely encased the retromandibular vein and inferiorly invaded branches of the external carotid artery. The FNA study was significant for a high-grade neoplastic lesion. These findings prompted immediate referral to the head-and-neck oncologist. The patient underwent a right-sided complete parotidectomy and the resected tumour was diagnosed as a salivary duct carcinoma. Following surgery, the patient completed 60 gray (Gy) of radiotherapy in 30 fractions. He had complete relief in pain-related symptoms following radiation therapy.

Case II

A 30-year-old female presented with a 3-month history of severe (10/10) pain in the left auricular and periauricular region. The pain began 7 months after she developed a generalised swelling over the left side of her face. The pain was electric, stinging and sharp in nature and would present either immediately following the first bite of a meal

or even at the thought of eating. The frequency and intensity of the pain episodes reduced during the course of the meal. Each episode of pain was associated with a feeling of cramping and sweating over the ipsilateral side of the face.

Her medical history was unremarkable. The examination of the head-and-neck region revealed the presence of a generalised swelling over the left side of her face which extended into the posterior mandibular fossa and limited (25 mm) her maximal mouth opening. A diagnosis was made and the patient was referred for magnetic resonance imaging (MRI) of the head-and-neck region and an incisional biopsy of the swelling was obtained.

The MRI study revealed the presence of a 4.2 cm × 1.6 cm lesion which had a hyperintense signal on T2 and a hypointense signal on T1 (with post-contrast enhancement). The lesion extended medially into the insertion of the pterygoid muscle and internal carotid artery and laterally into the masseter muscle. The incisional biopsy was positive for Grade I adenoid cystic carcinoma with perineural invasion. These findings prompted immediate referral to a head-and-neck oncologist. The tumour was deemed unresectable and she was referred for 70 Gy of radiotherapy in 33 fractions. She had complete relief in pain-related symptoms following radiation therapy.

Diagnosis and Management

Case I

In this case, the patient had severe episodic electric shock-like lancinating pain localised over the right side preauricular and posterior mandibular regions, which was triggered following the first bite of a meal. These episodes would continue for a few bites and subsequently subside. The patient did not have any episodes of electric shock-like pain until the next meal. In addition, the patient had a continuous background dull ache in the region. A few months following the onset of pain, the patient developed ipsilateral swelling over the right side posterior mandibular area and weakness in movements of the right lower lip. The patient did not have any pain with jaw functional or parafunctional movements, temporomandibular joint (TMJ) dysfunction or an abnormal dental examination. These findings suggested this to be a case of FBS.

The patient was prescribed carbamazepine 200 mg (3 times a day), lamotrigine 25 mg (3 times a day) and nortriptyline 25 mg (once at night). This resulted in resolution of the continuous dull ache. However, the patient had minimal relief in electric shock-like pain. In subsequent follow-up visits, the patient was placed on various combinations and dosages of medications, including paracetamol 1000 mg (3 times a day), ibuprofen 400 mg (3 times a day), celecoxib 100 mg (2 times a day) and gabapentin 300 mg (3 times a day). The patient had the most relief (mild-moderate) in electric shock-like pain with paracetamol 1000 mg (3 times a day), celecoxib 100 mg (2 times a day), gabapentin 300 mg (3 times a day) and carbamazepine 200 mg (3 times a day) combination regimen. The patient had complete relief in electric shock-like symptoms following radiotherapy.

Case II

In this particular case, the patient had a history of severe electric, stinging and sharp pain in the left auricular and periauricular regions that were associated with episodes of cramping and sweating in the ipsilateral region. Her episodes of pain triggered when she thought about eating something or after the first bite of a meal and they would subside following few bites of the meal. There were no reports of additional sensory, motor or autonomic symptoms. The patient did not have any pain with jaw functional or parafunctional movements. There were no signs or symptoms of odontogenic pathology or TMJ dysfunction. These findings suggested her to have FBS.

The patient was initially placed on tramadol 100 mg (3 times a day), amitriptyline 25 mg (once at night), gabapentin 100 mg (3 times a day), oxcarbazepine 300 mg (3 times a day) and celecoxib 200 mg (2 times a day). However, she had minimal relief in

symptoms. Over the next follow-up visits, the patient was placed on various medication combinations including lamotrigine 25 mg (3 times a day), pregabalin 100 mg (2 times a day), morphine 20 mg (2 times a day) and hyoscine 20 mg (3 times a day). The patient had moderate relief in symptoms with the combination regimen consisting of morphine 20 mg (2 times a day), pregabalin 100 mg (2 times a day), oxcarbazepine 300 mg (2 times a day) and hyoscine 20 mg (3 times a day). Nonetheless, she had complete relief in symptoms following radiotherapy.

Discussion

FBS is characterised by the presence of electric shock-like pain that occurs with the first bite of the meal. The pain is localised around the periauricular region and often associated with the feeling of cramping in the region. These symptoms eventually fade away during mastication, only to restart at the beginning of the next meal.[1-3] These symptoms were demonstrated in both of the cases. In general, patients with FBS have a history of trauma in the head-and-neck region or surgery involving sympathetic chain, parotid gland, parapharyngeal space, infratemporal fossa, carotid bifurcation or internal carotid artery. However, in recent years, few pre-operative cases without a history of trauma have also been reported.[6-9]

The pathophysiology of FBS is unknown. The most widely accepted theory suggests that FBS takes place secondary to the damage of the cervical sympathetic trunk or the postganglionic sympathetic efferent fibres which innervate the parotid gland. This may lead to the denervation of the sympathetic receptors located on the parotid myoepithelial cells. Concurrently, these cells then become sensitive to parasympathetic stimulation, which may result in intense supramaximal contractile response at the first bite.[3,14] This proposed mechanism may also explain the pre-operative occurrence of FBS. In Case I, the tumour had invaded the external carotid artery which may have had damaged the post-ganglionic efferent branches of sympathetic nerves and caused sympathetic denervation of the parotid gland. Similarly, in Case II, the tumour had encased the internal carotid artery which could have damaged the pre-ganglionic sympathetic trunk and resulted in dysfunction of sympathetic-parasympathetic system. This notion was clinically supported by the concurrent occurrence of sweating with pain in the ipsilateral side of the face.

These case reports highlight the importance of a thorough history and clinicoradiological examination of patients with pre-operative FBS without a history of trauma or surgery. In this case, the clinical symptoms could have been mistaken for trigeminal neuralgia, odontogenic issue or TMJ dysfunction. However, the association of symptoms with the first bite of the meal, absence of any dental pathology, lack of modification of pain with jaw functional and parafunctional movements and normal clinical examination findings suggested FBS. Furthermore, these cases emphasise the significance of a close follow-up of patients that present with pre-operative FBS because in both of these cases, pain preceded any other clinical sign or symptom associated with the presence of parotid gland tumour. Patients with uncommon signs and symptoms should undergo a thorough and meticulous series of investigations for rare and potentially fatal causes of pain.

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Authorship Contributions

Conceived and designed the analysis: KSN; Collected the data: KSN and RH; Contributed data or analysis tools: RH and AJ; Wrote the paper: KSN, RH and AJ.